



Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

Parathyroid adenoma in a young female presenting as recurrent acute pancreatitis with a brown tumour of the mandible—A case study

A.R. Kunte^{a,*}, V.S. Dube^b, S.S. Balwantkar^b, K.K. Kulkarni^b^a General Surgical residency, B. J. Medical College and Sassoon General Hospitals, Pune, India^b B. J. Medical College and Sassoon General Hospitals, Pune, India

ARTICLE INFO

Article history:

Received 11 September 2014

Accepted 13 November 2014

Available online 27 November 2014

Keywords:

Parathyroid adenoma

Brown tumour

Acute pancreatitis

Young female

ABSTRACT

INTRODUCTION: Primary hyperparathyroidism is usually seen in females above the age of 50 years, with a prevalence of 21/1000,¹ whereas the incidence in patients aged 12–28 years is less than 5%.² A solitary adenoma is responsible for 80% of cases of primary hyperparathyroidism.³ Primary hyperparathyroidism is most commonly asymptomatic.⁴ The incidence of acute pancreatitis associated with hyperparathyroidism is less than 10%.^{5,6} The incidence of hyperparathyroidism associated with a Brown tumour is less than 5%.⁷

PRESENTATION OF CASE: A 19 year old female patient presented with recurrent acute pancreatitis and swelling over the mandible. Complete investigative workup revealed a solitary parathyroid adenoma causing hyperparathyroidism. Surgical exploration with excision of the parathyroid adenoma was performed, following which the patient recovered uneventfully.

DISCUSSION: The patient was initially managed as a case of acute pancreatitis, and although not suspected initially, a high index of suspicion for hyperparathyroidism developed after a biopsy of the mandibular swelling showed the presence of osteoclastic giant cells indicating the possibility of a Brown tumour. Further investigations then revealed the presence of a solitary parathyroid adenoma with coexistent hyperparathyroidism which was then managed surgically.

CONCLUSION: The young age of the patient, and her presentation with acute pancreatitis and a Brown tumour of the mandible make this an extremely rare presentation of parathyroid adenoma.

© 2014 The Authors. Published by Elsevier Ltd. on behalf of Surgical Associates Ltd. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/3.0/>).

1. Case report

The patient in concern was a 19 year old female who presented with the complaints of acute onset pain in abdomen. The pain was characteristic of acute pancreatitis. The patient was three months postpartum at the time of presentation having undergone a caesarean section in her 7th month of pregnancy. She also had two prior episodes of acute pancreatitis during her pregnancy: once during her third month and the second during her 7th month. Both episodes were managed conservatively and resolved without complications.

She also frequently complained of back pain in the thoracic region and deep seated bony pain in the right hip and thigh.

She noticed a painless swelling on the left side of her jaw five months back, which appeared insidiously and gradually increased in size to its current size of around 4 × 4 cm.

There was no history of prior neck irradiation, neck surgery or radio-isotope administration. She had neither suffered from renal calculus disease nor been incidentally diagnosed with it in the past.

On examination, her findings were consistent with that of acute pancreatitis, with tachycardia and abdominal tenderness which was maximal in the epigastric region.

There was a globular, painless, non-tender, non-pulsatile swelling 4 × 4 cm in size situated over the ramus of the left side of the mandible. This swelling was hard in consistency and fixed to the underlying mandible. The swelling was not visible intra-orally. There were no signs of inflammation or sinus formation in the skin overlying and around the swelling. The swelling was free from the overlying skin. Neck examination revealed no abnormal findings or lymphadenopathy.

2. Radiological investigations

- 1) *USG abdomen:* An abdominal USG done at the time of admission showed changes consistent with acute pancreatitis.
- 2) *CT scan – abdomen:* CT scan of the abdomen confirmed the diagnosis of acute pancreatitis. There was also the presence of a 5 × 4.9 × 4.7 cm sized pseudocyst in the tail region of the

* Corresponding author. Tel.: +91 8806664737.

E-mail address: a.kunte13@gmail.com (A.R. Kunte).



Fig. 1. Swelling present on the ramus of the left side of the mandible.

pancreas, with a wall thickness of 3 mm. There was no evidence of renal calculus disease, adrenal masses, pancreatic calcifications or pancreatic duct calculi.

- 3) *USG neck*: 2.4×1.2 cm sized hypoechoic mass was seen at inferior pole of the right side of the thyroid gland, suggestive of a parathyroid adenoma.
- 4) *CT scan neck*: 1.8×1.2 cm sized heterogeneously enhancing mass was seen on right postero-medial border of the thyroid gland near the inferior pole suggestive of a parathyroid adenoma. Lytic lesions with soft tissue components were noted on

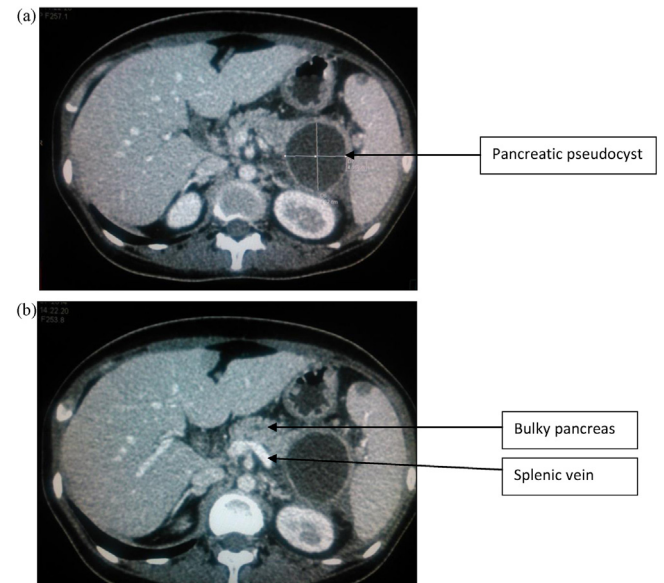


Fig. 2. (a) CT scan section demonstrating the pseudocyst in the pancreas. (b) CT scan section demonstrating features of acute pancreatitis.

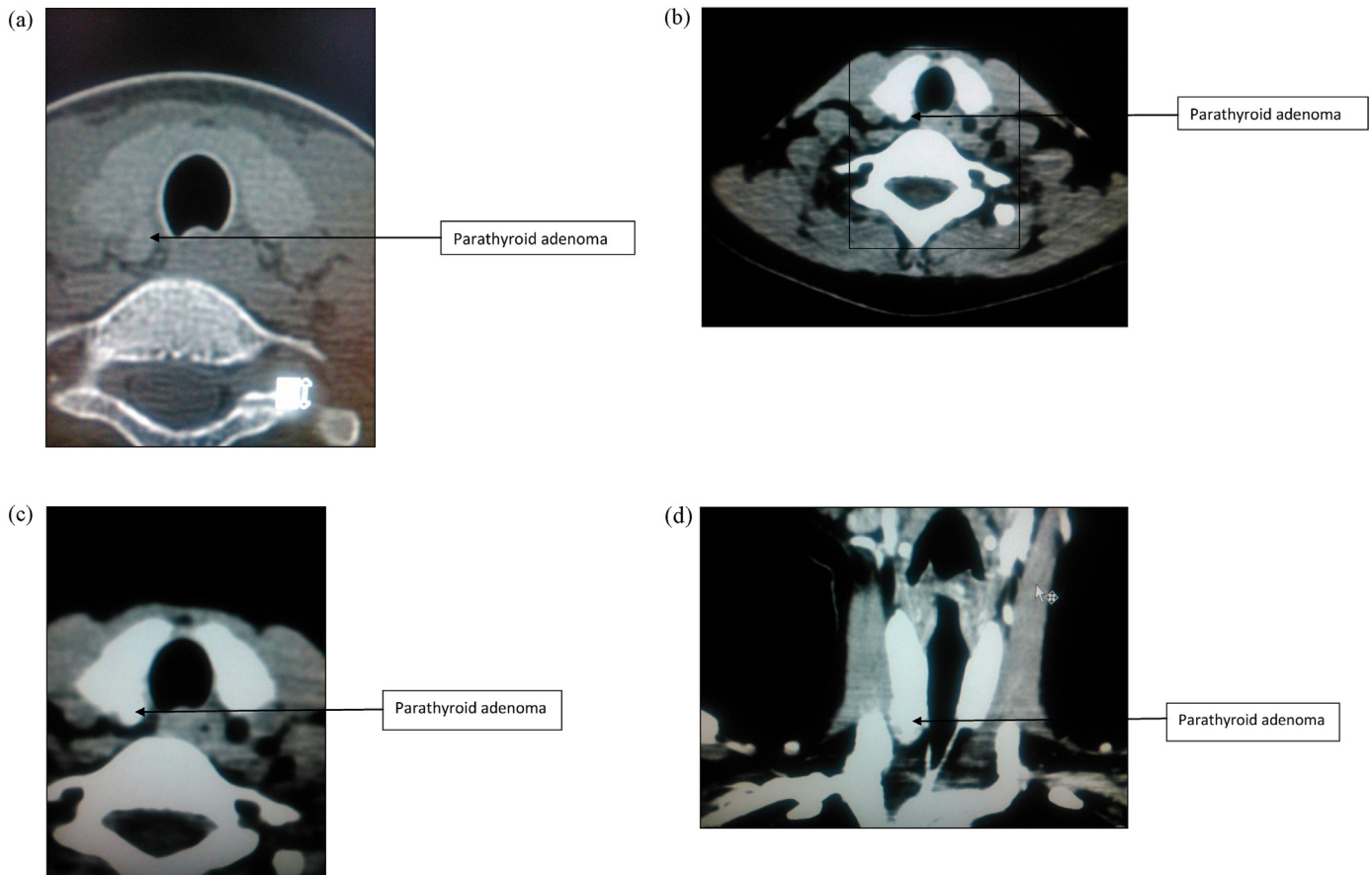


Fig. 3. (a) Plain CT scan section demonstrating a parathyroid adenoma on the right side. (b) Contrast enhanced CT scan section demonstrating a well enhancing parathyroid adenoma on the right side. (c) Magnification of Fig. 3b demonstrating a well enhancing parathyroid adenoma on the right side. (d) Coronal section of contrast enhanced CT scan demonstrating a parathyroid adenoma on the right side at the inferior pole.

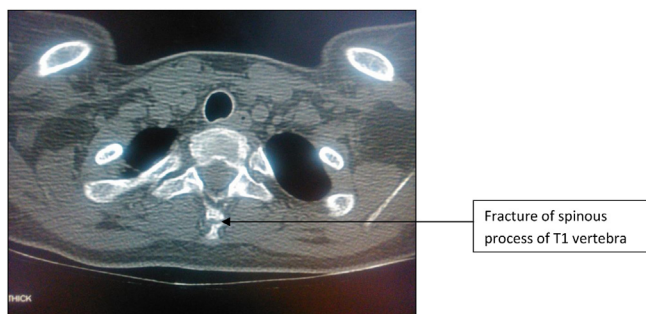


Fig. 4. CT scan demonstrating a pathological fracture of the spinous process of T1 vertebra.

ramus of mandible on the left side, measuring 2.8×2.3 cm, suggestive of a Brown tumour. Fracture of spinous process of the T1 vertebra was also detected.

- 5) *Parathyroid Tc99m-Sestamibi scan*: Well defined parathyroid adenoma in the region of the lower pole on the right side of the thyroid gland. No evidence of ectopic parathyroid tissue was found.

3. Laboratory investigations

All routine laboratory investigations – Hemogram, Liver function tests, Renal function tests and Sr. Electrolytes were done and were found to be within normal limits.

- 1) *Sr. Amylase*: 30 IU/L (0.0–40.0 IU/L) on admission
- 2) *Sr. Lipase*: 28 U/L (50–200 U/L) on admission
- 3) *Sr. Calcium*: 11.5 mg% (9–11 mg%) pre-operatively
- 4) *Sr. PTH*: 908.90 pg/mL (15–65 pg/mL) pre-operatively
- 5) *Sr. Alkaline phosphatase*: 1782 U/L (64–306 U/L) pre-operatively

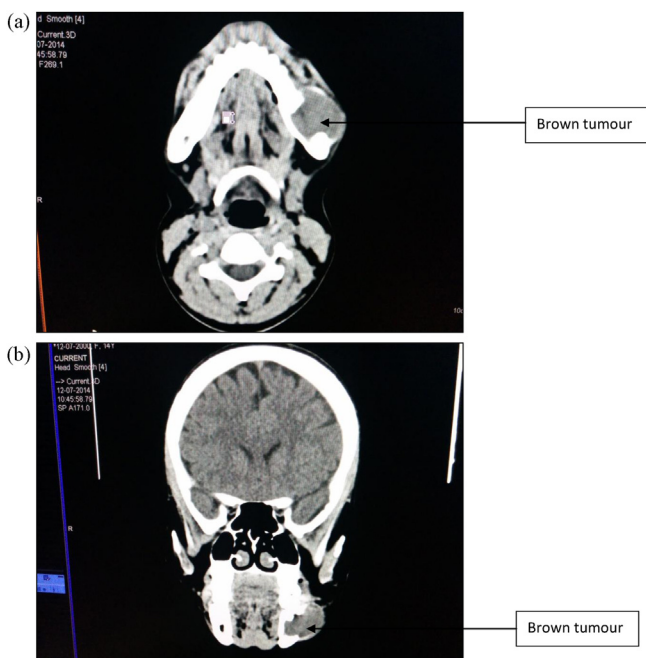


Fig. 5. (a) CT scan demonstrating a Brown tumour of the left side of the mandible. (b) CT scan coronal section demonstrating a Brown tumour of the left side of the mandible.

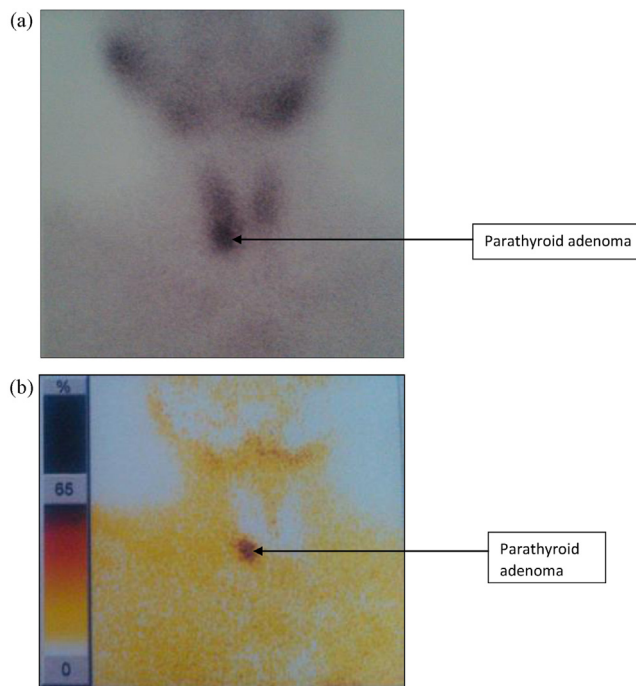


Fig. 6. (a) Parathyroid Tc99m-sestamibi scan demonstrating a parathyroid adenoma of the right inferior parathyroid gland. (b) Subtraction images of the parathyroid scan demonstrating a well defined parathyroid adenoma of the right inferior parathyroid gland.

4. Tissue diagnosis

Biopsy of the mandibular swelling was done which revealed the presence of osteoclastic giant cells, which when correlated with

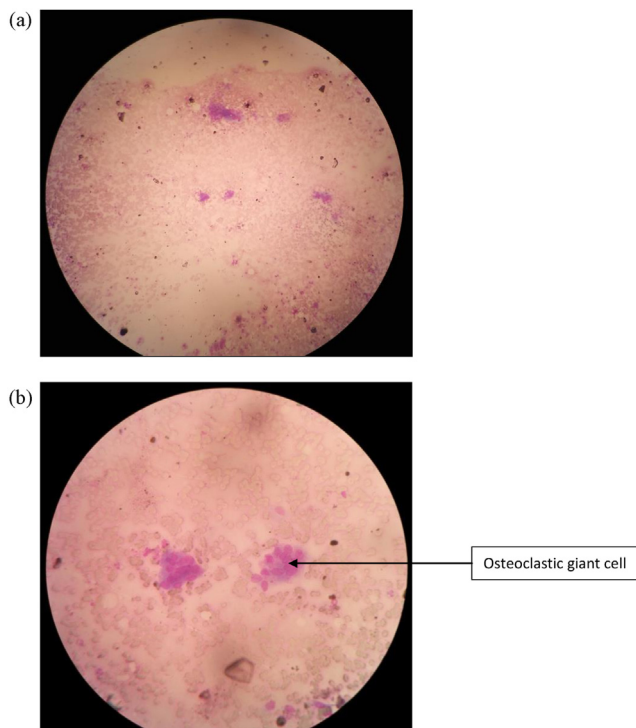


Fig. 7. (a) Low power view of the biopsy of the mandibular swelling showing the presence of numerous osteoclastic giant cells. (b) High power view of the biopsy of the mandibular swelling focusing on the osteoclastic giant cells.

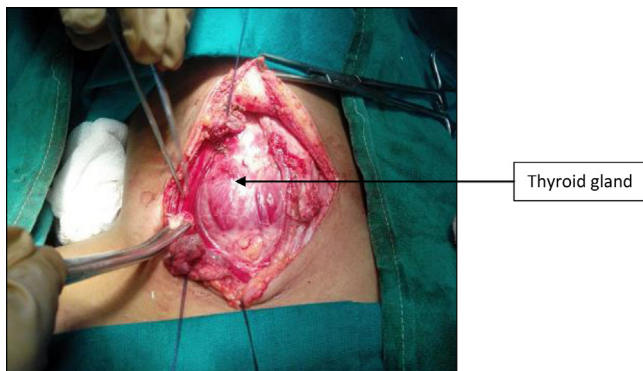


Fig. 8. Strap muscles divided on the right to gain exposure to the underlying thyroid gland.

the clinical and radiological findings, confirmed the diagnosis of a Brown tumour of hyperparathyroidism.

5. Treatment

The patient was electively posted for surgery. Excision of the parathyroid adenoma with exploration of the remaining parathyroid glands was done.

Neck was accessed via a transverse skin crease incision. Subplatysmal flaps were raised and strap muscles retracted laterally. Strap muscles of the right side were divided horizontally to expose the right lobe of the thyroid.

The thyroid was retracted medially to expose the posteromedial border, thus enabling visualization of the parathyroid adenoma. It was seen as a smooth, oval, yellowish structure near the lower pole of the right lobe of the thyroid, seen to be arising from the right inferior parathyroid gland, measuring approximately 2×1 cm in size.

The adenoma was then dissected free from the surrounding thyroid gland, finally isolating its blood supply. It was then excised

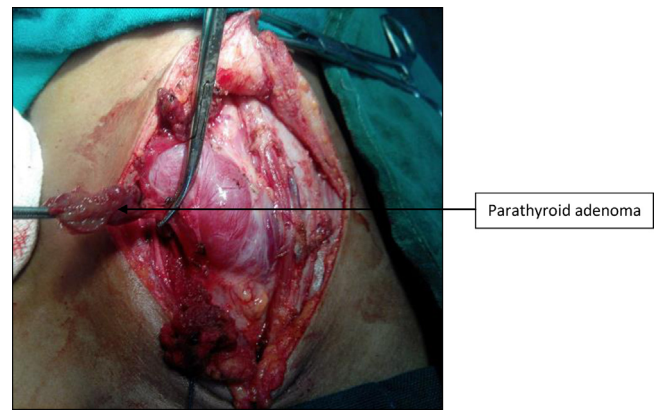


Fig. 10. Blood vessel supplying the parathyroid adenoma is clamped, ligated, and the adenoma excised.

after ligating the feeding vessel, and sent for frozen section. Frozen section report confirmed the mass to be a parathyroid adenoma.

The rest of the parathyroid glands were explored and were found to be normal, hence were left in situ.

A negative suction drain was placed below the strap muscles and the incision closed in layers over it. Skin was closed with subcuticular sutures. The sutures healed without any post-operative complications.

6. Histopathology

The excised parathyroid adenoma was then sent for histopathological examination. The diagnosis of parathyroid adenoma was confirmed on histopathological examination by presence of normal parathyroid glandular tissue, markedly reduced amount of adipose tissue and an intact capsule.

7. Post-operative recovery

In the post-operative period, the patient was administered a calcium gluconate drip continuously for the first 3 post-operative days. The patient developed 3 separate episodes of tetany on post-operative day 3, 5 and 6 all of which were managed by administering a bolus dose of IV calcium gluconate.

Oral administration of Calcium gluconate and Vit. D3 was started on the 2nd post-operative day.

The negative suction drain was removed on the 3rd post-operative day.

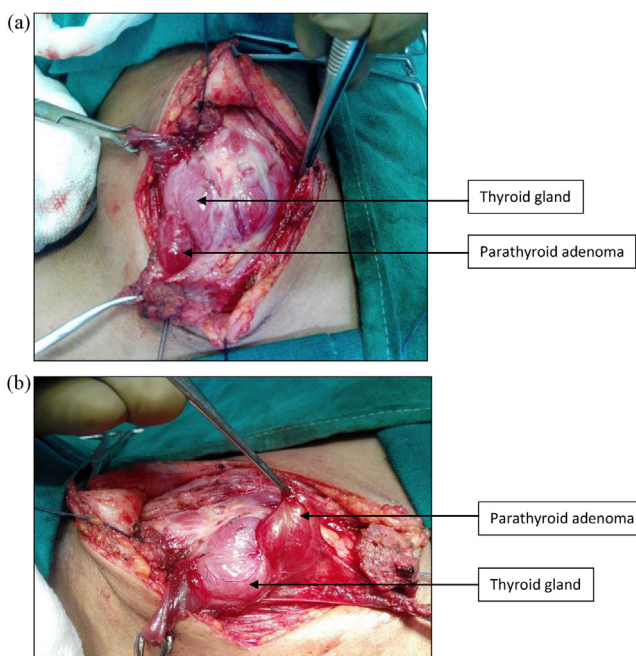


Fig. 9. (a) Thyroid gland retracted medially to expose the parathyroid adenoma. (b) Lateral view of the parathyroid adenoma.



Fig. 11. Excised parathyroid adenoma.

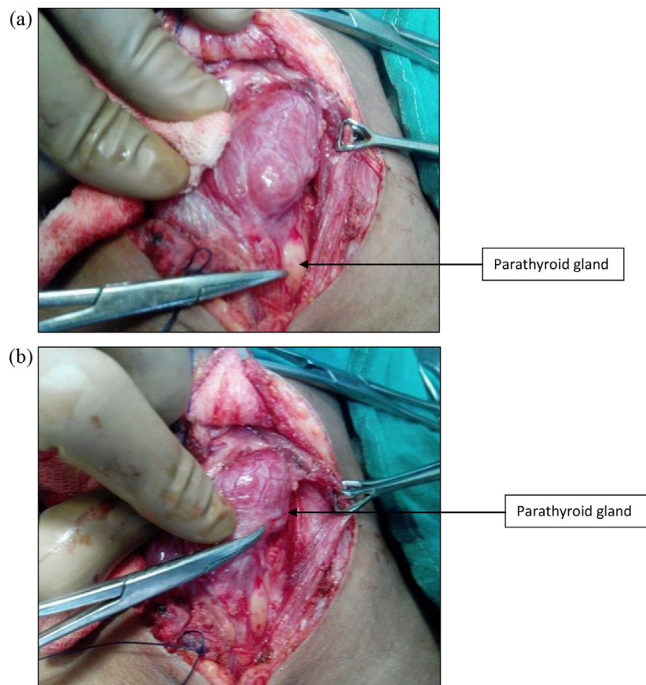


Fig. 12. (a) Left thyroid lobe retracted medially to expose normal left inferior parathyroid gland. (b) Left lobe of thyroid gland retracted medially to expose normal left superior parathyroid gland.

The post-operative laboratory investigations yielded the following results:

- 1) Sr. Calcium: 10.5 mg% (9–11 mg%)
- 2) Sr. PTH: 9.24 pg/mL (15–65 pg/mL)
- 3) Alkaline phosphatase: 1696 U/L (64–306 U/L)

The rest of the routine investigations were within normal limits. The sutures healed without any complications and were removed on the 10th post-operative day.

The patient was discharged on the 12th post-operative day after full recovery (Figs. 1–14).



Fig. 13. Healed surgical scar of the patient on the 12th post-operative day.



Fig. 14. Slide photograph of the parathyroid adenoma.

Conflict of interest

None.

Funding

Funding from external sources was not necessary since the patient in concern was treated in our institute, which is a public sector hospital, and all the treatment to the patient was provided free of cost.

Ethical approval

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A special consent was also taken for the release of three actual photographs of the patient for the purpose of this case report.

Author contributions

Dr. Aditya R. Kunte: Diagnosis, Operative procedure, Writing the case report. Dr. Vandana S. Dube: Diagnosis, Operative procedure, Review of the written case report. Dr. Sachin S. Balwantkar: Diagnosis, Operative procedure. Dr. Kalpana S. Kulkarni: Histopathological diagnosis.

References

1. Adami S, Marcocci C, Gatti D. Epidemiology of primary hyperparathyroidism in Europe. *J Bone Miner Res: Off J Am Soc Bone Miner Res* 2002;**17**(2): N18–23.
2. Yeh Michael W, Ituarte Philip HG, Zou Hui Cynthia, Nishimoto Stacie, Liu In-Lu Amy, Harari Avital, et al. Incidence and prevalence of primary hyperparathyroidism in a racially mixed population. *J Clin Endocrinol Metab* 2013;**98**: 1122–9.
3. Udelsman R. Six hundred fifty-six consecutive explorations for primary hyperparathyroidism. *Ann Surg* 2002;**235**:665–70, discussion 70–2.
4. Zhao Lin, Liu Jian-min, He Xiao-Yan, Zhao Hong-yan, Sun Li-hao, Tao Bei, et al. The changing clinical patterns of primary hyperparathyroidism in Chinese patients: data from 2000 to 2010 in a single clinical center. *J Clin Endocrinol Metab* 2013;**98**(February (2)).
5. Lee CC, Chao AS, Chang YL, Peng HH, Wang TH, Chao A. Acute pancreatitis secondary to primary hyperparathyroidism in a postpartum patient: a case

- report and literature review. *Taiwan J Obstet Gynecol* 2014;**53**(June (2)):252–5, <http://dx.doi.org/10.1016/j.tjog.2013.01.029>.
6. Biondi A, Persiani R, Marchese M, Cananzi F, D'Ugo D. Acute pancreatitis associated with primary hyperparathyroidism. *Updates Surg* 2011;**63**(June (2)):135–8.
7. Chowdhury S, Aggarwal A, Mittal N, Shah A. Brown tumor of hyperparathyroidism involving craniomaxillofacial region: a rare case report and literature review. *Minerva Stomatol* 2013;**62**(September (9)): 343–8.

Open Access

This article is published Open Access at scimedirect.com. It is distributed under the [IJSCR Supplemental terms and conditions](#), which permits unrestricted non commercial use, distribution, and reproduction in any medium, provided the original authors and source are credited.